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## ORIGINAL ARTICLE

# The influence of maternal vulnerability and parenting stress on chronic pain in adolescents in a general population sample: The TRAILS study

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## Abstract

Investigating possible psychosocial predictors of unexplained chronic pain in adolescents is crucial in understanding its development and prevention. A general population sample of adolescents ( $n = 2230$ ) from the TRAILS cohort study was investigated longitudinally to assess the influence of maternal vulnerability, in terms of anxiety, depression and stress, and parenting stress at age 10–12 years, on the presence of chronic pain at age 12–15 years. Of these adolescents, 269 (12.9%) reported experiencing chronic pain, of which 77% reported severe chronic pain and 22% reported multiple chronic pain. Maternal anxiety, maternal stress and higher levels of parenting stress were related to chronic pain at a later age. Subgroup analyses showed similar results for adolescents with severe chronic pain. Mediation analyses indicated that parenting stress mediates the effect between maternal anxiety, or stress, and chronic pain. The findings suggest that interventions to diminish maternal feelings of anxiety and stress, while in turn adjusting maternal behaviour, may prevent the development of chronic pain in adolescence.

## 1. Introduction

Chronic pain in children and adolescents is widespread, with prevalences of up to 25% (Perquin et al., 2000; Huguet and Miro, 2008; Stanford et al., 2008). A majority of the children report pain in the absence of an underlying physical condition.

The role of mothers in child chronic pain has been investigated in the past (Liakopolou-Kairis et al., 2002). Maternal psychological vulnerability, in this context defined as the general tendency to react with anxiety and depression in stressful situations, has been a focus of research. Cross-sectional studies investigating children with recurrent abdominal pain (RAP) found that mothers had higher levels of anxiety, depression and somatisation. (Garber et al., 1990;

Walker and Greene, 1989). These studies were conducted in clinical samples and may not be representative of the general population. However, evidence for a relationship between maternal vulnerability and abdominal pain was also found in longitudinal studies in general population samples. Higher levels of neuroticism in mothers with children with abdominal pain were found (Hotopf et al., 1998), as were higher levels of anxiety and depression in mothers of children with abdominal pain (Ramchandani et al., 2006; Mortimer et al., 1992).

The question that arises from these studies is how aspects of maternal vulnerability influence child chronic pain. Social learning theory (Bandura, 1977; Craig, 1983) as a model for understanding the influence of maternal vulnerability, proposes modeling and

reinforcement as possible mechanism (Levy et al., 2007). Children with pain have been found to model parents' physical symptoms or their attention to symptoms (Osborne et al., 1989; Rickard, 1988; Walker et al., 2006). In addition, reinforcement of pain may occur when anxious parents give their child more attention, as was shown in studies on symptoms (Walker et al., 1993; Whitehead et al., 1994).

A large body of research on parental mental health and child development has shown that psychological problems can compromise parenting abilities. Abidin presents a model, whereby parent characteristics, such as the presence of anxiety or depression, among others influence the levels of parenting stress experienced, which subsequently influences parenting behavior (Abidin, 1992). The influence of parenting stress has only previously been investigated as a consequence in pediatric pain (Eccleston et al., 2004), rather than a determinant. While the direct relationship of maternal vulnerability and chronic pain has been assessed, the possibility of this relationship actually being mediated by parenting stress has not previously been explored.

The present study aims to investigate the influence of maternal vulnerability at baseline (child age 10–12 years) on the presence of chronic pain in adolescents at age 12–15 years. In the current study this is operationalised by investigating the influence of maternal anxiety, stress, depression and parenting stress as determinants. In addition, the study set out to investigate these factors in a community sample, as the outcomes for children in clinical settings cannot be generalized to all children with chronic pain as they tend to be more severe (Huguet and Miro, 2008). In addition, studying adolescents with chronic pain in the community may provide information which will aid in early identification of children with chronic pain.

## 2. Methods

### 2.1 Participants

Adolescents were all part of the Tracking Adolescents' Individual Lives Survey (TRAILS), a general population study following Dutch preadolescents from age 10. TRAILS is designed to chart and explain the development of mental and social development from preadolescence into adulthood. Sample selection involved two steps. First, selected municipalities from the North of the Netherlands were requested to give names and addresses of all inhabitants born between 01-10-1989 and 30-09-1990 (first two municipalities) or 01-10-1990 and 30-09-1991 (last three municipali-

ties), yielding 3483 names. Simultaneously, primary schools (including schools for special education) within these municipalities were approached with the request to participate in TRAILS; i.e., pass on students' lists, provide information about the children's behavior and performance at school, and allow class administration of questionnaires and individual testing (neurocognitive, intelligence, and physical) at school. School participation was a prerequisite for eligible children and their parents to be approached by the TRAILS staff, with the exception of children already attending secondary schools (<1%), who were contacted without involving their schools. Of the 135 primary schools within the municipalities, 122 (90.4% of the schools accommodating 90.3% of the children) agreed to participate in the study. If schools agreed to participate, parents (or guardians) received two brochures, one for themselves and one for their children, with information about the study; and a TRAILS staff member visited the school to inform eligible children about the study. Shortly thereafter a TRAILS interviewer contacted parents by telephone to provide additional information, answer questions, and ask whether they and their son or daughter were willing to participate in the study. Respondents with an unlisted telephone number were requested by post to provide their telephone number. If they did not respond to that letter, nor to a reminder sent a few weeks later, staff members paid personal visits to their house. Parents who refused to participate were asked for permission to call back in about two months to minimize the number of refusals due to temporary reasons. If both parents and children agreed to participate, parental written informed consent was obtained after the procedures had been fully explained. Children were excluded from the study if they were incapable of participating due to mental retardation or a serious physical illness or handicap, or if no Dutch-speaking parent or parent surrogate was available, and it was not feasible to administer part of the measurements in the parent's language. Of all children approached for enrollment in the study (i.e., selected by the municipalities and attending a school that was willing to participate,  $n = 3145$ ), 6.7% were excluded because of incapability or language problems. Of the remaining 2935 children, 76.0% ( $n = 2230$ ) were enrolled in the study (i.e., both child and parent agreed to participate). Responders and non-responders did not differ with respect to the prevalence of teacher-rated problem behavior. Furthermore, no differences between responders and nonresponders were found regarding associations between sociodemographic variables and mental

health outcomes (De Winter et al., 2005). The North of the Netherlands has relatively few immigrants: only 10.6% of the children originated from a non-western country (mostly Morocco, Turkey, Surinam, the Dutch Antilles, and Indonesia). The percentage of children that has lived with the same parents from birth to preadolescence is 76.3. The 23.7% for whom it was not the case can be divided into children who have always lived with a single parent (2.4%), children who experienced a divorce and have lived with a single parent since then (12.9%), and children who experienced a divorce and have lived with a stepparent since then (8.4%). The mean age of mothers was 40.45 years (SD 4.70 years). Most families (78.0%) included two or three children. Socio-economic status (SES) was assessed from information on the mother and father's education and employment, as well as income. SES was low in 25.3%, intermediate in 49.5% and high in 25.2% of the cases.

### 2.1.1 Study procedure

Well-trained interviewers visited one of the parents or guardians (preferably the mother, 95.6%) at their homes to administer an interview covering a wide range of topics, including the child's developmental history and somatic health, parental psychopathology and care utilization. In addition to the interview, the parent was asked to complete a self-report questionnaire. Children were measured at school, where they completed questionnaires, in groups, under the supervision of one or more TRAILS assistants. In addition to that, information processing capacities (neurocognitive tasks), intelligence, and a number of biological parameters (including weight and height) were assessed individually (at school, except for saliva samples, which were collected at home). Teachers were asked to complete a brief questionnaire for all TRAILS-children in their class. Of all the children approached ( $n = 3145$ ), 6.7% ( $n = 211$ ) were excluded because of mental or physical incapability or language problems, leaving a total of 2934. Finally, 76.0% participated in the baseline assessment (T1:  $n = 2230$ , mean age 11.1 years, SD 0.56, range 10.0–12.0), and 96.4% ( $n = 2149$ ) were assessed at follow-up (T2: mean age 13.6 years, SD 0.53, range 12.0–15.0), held approximately 2.2 years after T1 (mean follow-up time 2.47, SD 0.48, range 0.73–3.25). The T1 assessment was conducted from March 2001 to July 2002, and the T2 assessment was conducted from September 2003 to December 2004. The current study presents data from the first and second measurement waves of TRAILS, running from March 2001 to July 2002 and

September 2003 to December 2004, respectively. At wave 1 (T1) 2230 children aged 10–12 years were enrolled in the study and at wave 2 (T2) 2149 adolescents aged 12–15 years participated in the study.

Children and parents completed questionnaires biennially. More detailed information on the cohort can be found in De Winter et al. (2005) and Huisman et al. (2008). In the current study determinants (maternal anxiety, depression, stress, and parenting stress) were measured at T1 and the outcome measure chronic pain was measured at T2. All child measures and maternal measures were self-reported.

## 3. Measures

### 3.1 Determinants

#### 3.1.1 Mother

##### 3.1.1.1 Maternal vulnerability at T1

The Depression Anxiety Stress Scales 21 (DASS-21) was used to measure depression, anxiety and stress and is derived from the original 42-item self-report measure of depression, anxiety, and stress (DASS; Lovibond and Lovibond, 1995). The DASS-21 consists of 21 items, which are rated on a four-point severity scale, measuring experiences over the past week. Examples of items are "I found it hard to wind down" (stress), "I was worried about situations in which I might panic and make a fool of myself" (anxiety) and "I couldn't seem to experience any positive feeling at all" (depression). Internal consistencies of the three scales were 0.83 for depression, 0.78 for anxiety, and 0.86 for stress. The factor structure of the DASS has been replicated using the DASS-21 in a large population sample of adults in the United Kingdom (Henry and Crawford, 2005).

##### 3.1.1.2 Parenting stress at T1

This variable was measured by the Dutch short-version of the Parental Stress Index (PSI; Abidin, 1983; De Brock et al., 1992). The measure contains 25 statements, in the parent and child domain, which can be answered on a sixpoint scale ranging from completely disagree (1) to completely agree (6). Parent domains are: sense of competence, attachment, depression, and parental health. Child domains are: adaptability, mood, distractibility/hyperactivity, demandingness, reinforces parent, and acceptability. Example of items are "I feel trapped by my responsibilities as a parent", and "There are some things my child does that really

bother me a lot". This version of the PSI has shown excellent internal consistency (Cronbach's  $\alpha = 0.92$ – $0.95$ ), and differentiates between clinical and non-clinical groups (De Brock et al., 1992). Internal consistency in our sample was also high (parent domain 0.86; child domain 0.92). Since parent and child domains were highly correlated ( $r = 0.78$ ), a total parenting stress score was calculated by summing all items (Cronbach's  $\alpha = 0.94$ ), which was used in the analyses.

### 3.1.2 Child

#### 3.1.2.1 Pain at T1

Adolescents were asked questions regarding pain at T1. Adolescents were asked whether they had suffered from earache, back or neckpain, headache or migraine and abdominal pain, being the most common pain locations. Questions could be answered as not (1), a little (2) quite a lot (3), and a lot (4). This information was used to give an indication of the amount of pain at baseline in the final analyses. A dummy variable was created, whereby adolescents who indicated experiencing quite a lot (3) or a lot of pain (4) on any of the pain questions were classified as having pain. Unfortunately these questions cannot be used to assess whether children experience chronic pain, because an indication of duration is lacking, which is needed to assess chronic pain. At T2 the Painlist was used, which has been used and published extensively in previous studies (Perquin et al., 2000; Hunfeld et al., 2001; Merlijn et al., 2003). Information on duration is used to assess whether children experience pain longer than three months, as assessed by the Painlist which was used at T2.

### 3.2 Outcome measure

#### 3.2.1 Chronic pain at T2

Questions on pain were based on the Painlist (Perquin et al., 2000). Respondents indicate (a) if they have experienced a certain pain, (b) pain intensity rated on a scale from 0 (no pain) to 10 (unbearable pain), (c) duration of pain in the last year, from less than 4 weeks to more than 3 months, and (d) frequency of experienced pain (less than once a month to almost every day). Questions were asked about earache, neck pain, headache or migraine, back pain, shoulder pain, arm pain, leg pain and abdominal pain. Chronic pain in children is defined as having pain for longer than three months, and severe chronic pain is defined as

having pain on a weekly basis, for at least three months, where the average pain intensity is  $>5$  on a scale of 0–10 (Perquin et al., 2000).

## 4. Data analysis

Prevalence of chronic pain, different pain sites, severe chronic pain and multiple chronic pain in the sample was calculated in terms of frequencies and percentages. Between the chronic pain group and the comparison group comparisons were made on sex and age and all the subscales of maternal determinants by means of logistic regressions. Medians and interquartile ranges are presented.

In order to investigate the relationship between maternal vulnerability and parenting stress on the one hand, and chronic pain on the other hand, regression analyses were carried out, controlling for child sex and pain at T1. These logistic regression analyses with chronic pain (T2) as the outcome measure were carried out, separately, for each of the maternal variables, i.e. anxiety, depression, stress and parenting stress. The logistic regression analyses were conducted separately for each determinant, as the determinants were correlated (Table 2).

In the final step of the analyses the mediational influence of parenting stress was investigated. This was done by following Baron and Kenny's (1986) definition of mediation. Here  $X$  is the determinant,  $Y$  the outcome variable and  $M$  the possible mediator variable. Mediation effect is present if (1)  $X$  significantly predicts  $Y$  (relationship  $c$ ), (2)  $X$  significantly predicts  $M$  (relationship  $a$ ) and  $M$  significantly predicts  $Y$  (relationship  $b$ ), and (3)  $M$  significantly predicts  $Y$  controlling for  $X$  (relationship  $c'$ ). Bootstrapping analyses were conducted according to methods described by Preacher and Hayes (2004), using their SPSS macro. Bootstrapping analysis is preferable as firstly, it does not rely on the assumption that the data present a normal distribution and secondly, it reduces the likelihood of Type I errors as the number of inferential tests are minimized (Preacher and Hayes, 2004). Analyses were run for those maternal variables that showed a significant relationship with child chronic pain, thereby fulfilling the first rule of mediation effects (i.e. significant relationship between  $X$  and  $Y$ ). In these analyses child chronic pain at T2 was entered as the dependent variable ( $Y$ ), maternal anxiety, stress or depression ( $X$ ) as the independent variables and parenting stress as the possible mediator ( $M$ ). Data will be presented in terms of total, direct and indirect effects. The total effect refers to path  $c$ , between  $X$  and  $Y$ , before mediation. The direct effect refers to path  $c'$



in the mediation model (between  $X$  and  $Y$  after  $M$  has been controlled). The indirect effect is the amount of mediation, and thus the reduction of the effect of  $X$  on  $Y$ , or  $c-c'$ .

Lastly, the same analysis as presented in the final model was run with 'severe chronic pain' as the outcome measure.

## 5. Results

### 5.1 Demographics and chronic pain

At T1 597 adolescents (28.4% of 2102 children, data missing for 128 children, total sample at T1 = 2230) reported having had 'quite a lot' or 'a lot' of pain, with 14.8% reporting abdominal pain, 12.2% reporting headaches, 8.7% back or neck pain and 6.4% earache. Girls were more likely to report pain at T1 (Chi square = 4.80,  $p = 0.028$ ) than boys, whereas the child's age did not influence the presence of pain at T1 ( $t = -0.38$ ,  $p = 0.70$ ). Pain at T2 was assessed in a sample of 2149 participants of which 2093 cases completed the pain questions, which means 2.6% ( $n = 56$ ) of data was missing. At T2 12.9% of adolescents ( $n = 269$ ) reported having chronic pain, 168 (62.5%) girls and 101 (37.5%) boys (Table 1). The pain locations reported can be broken down into the following: 32% headache or migraine, 25% back pain, 25% leg pain, 16% abdominal pain, 14% neck pain, 9% shoulder pain, 6% arm pain and 5% earache (these percentages do not add up to 100 since some adolescents had multiple chronic pain). Within these pain location groups there was a sex difference for abdominal pain with girls being more likely to experience abdominal pain ( $n = 35$ ) than boys ( $n = 7$ ; Chi-square = 9.25,  $p = 0.02$ ), and a similar trend was found for headache with more girls reporting headache (girls  $n = 62$ , boys  $n = 25$ ; Chi-square = 3.65,  $p = 0.056$ ). Of those adolescents with chronic pain 59 (22%) experienced multiple chronic pain, i.e. suffering from more than one pain in the last 3 months. Thus 2.8% of children in the overall cohort experienced multiple chronic pain. There was no sex difference between the proportion of adolescents with multiple chronic pain compared with that for single chronic pain. Severe chronic pain, which is defined as the weekly occurrence of pain lasting more than 3 months, with pain intensity above 5 on a scale of 0–10, was present in  $n = 208$  (9.9%) of all adolescents. This indicates that of the children with chronic pain, the majority ( $n = 208$ ; 77%) is likely to experience severe chronic pain. Girls were more likely to experience severe chronic

**Table 1** Number and percentages of chronic pain, and medians (interquartile ranges) for child and maternal determinants, for adolescents in the chronic pain group and the comparison group.

	Chronic pain group	Comparison group
<i>Child variables</i>		
Sex, girls (%)	168 (62.5)	905 (49.5)***
Age at T2, mean years (SD)	13.6 (.55)	13.6 (.52)
Pain at T1 <sup>a</sup> , $n$ (%)	597 (28.4)	1505 (71.6)
Chronic pain at T2 <sup>b</sup> , $n$ (%)	269 (12.9)	1824 (87.1)
Severe chronic pain at T2, $n$ (%)	208 (9.9)	1885 (90.1)
Multiple chronic pain at T2, $n$ (%)	59 (2.8)	2034 (97.2)
<i>Chronic pain locations at T2</i>		
Earache, $n$ (%)	13 (5)	
Neck pain, $n$ (%)	37 (14)	
Headache or migraine, $n$ (%)	87 (32)	
Back pain, $n$ (%)	68 (25)	
Shoulder pain, $n$ (%)	25 (9)	
Arm pain, $n$ (%)	15 (6)	
Leg pain, $n$ (%)	66 (25)	
Abdominal pain, $n$ (%)	42 (16)	
<i>Maternal vulnerability at T1</i>		
Stress	0.57 (0.57)	0.43 (0.57)**
Anxiety	0.14 (0.29)	0.00 (0.14)***
Depression	0.14 (0.43)	0.14 (0.43)
Parenting stress	1.68 (1.17)	1.52 (.88)***

Differences between the adolescents in the chronic pain group and adolescents in the comparison group.

\*\* $p < 0.01$ .

\*\*\* $p < 0.001$ .

<sup>a</sup>At T1  $n = 2230$ , with data missing for 128 for Pain at T1, therefore  $n = 2102$ .

<sup>b</sup>At T2  $n = 2149$ , with data missing for 56 children for Chronic pain at T2, therefore  $n = 2093$ .

pain ( $n = 140$ , 67%) than boys ( $n = 68$ , 33%, Chi-square = 9.22,  $p < 0.01$ ). The chronic pain group differed from the comparison group on maternal stress and anxiety, with mothers in the chronic pain group having higher scores, and also reporting higher levels of parenting stress (Table 1). Mothers in the chronic pain group did not differ significantly from mothers in the comparison group on depression. Correlations of the determinants are displayed in Table 2 and correlations were high between maternal depression, anxiety and stress.

**Table 2** Pearson correlations between determinants at T1.

	Maternal stress	Maternal depression	Parenting stress
Maternal anxiety	.58***	.58***	.25***
Maternal stress	–	.71***	.40***
Maternal depression		–	.31***

\*\*\* $p < 0.001$ .

**Table 3** Four separate logistic regressions, expressed in odds ratio's, with chronic pain at T2 as outcome measure and maternal anxiety, stress, depression and parenting stress (all at T1) as determinants. The same regressions are presented with severe chronic pain at T2 as the outcome measure.

Independent variables	Chronic pain			Severe chronic pain		
	<i>p</i>	OR <sup>a</sup>	95% CI	<i>p</i>	OR <sup>a</sup>	95% CI
<i>Maternal vulnerability</i>						
Anxiety	.009	1.71	1.14–2.56	.005	1.85	1.20–2.83
Stress	.005	1.52	1.13–2.04	.006	1.57	1.40–2.17
Depression	.350	1.20	.82–1.77	.129	1.38	.91–2.09
<i>Parenting</i>						
Parenting stress	.000	1.45	1.23–1.72	.000	1.47	1.22–1.77

<sup>a</sup>All analyses are adjusted for child sex and pain at T1.

## 5.2 Determinants of chronic pain

The separate logistic regressions show that maternal stress (OR = 1.52; CI 95% 1.13; 2.04) and anxiety (OR = 1.71, CI 95% 1.14; 2.56) were both positively related to the presence of chronic pain (Table 3), with higher levels of maternal stress or anxiety being related to the presence of chronic pain in the child. Increased levels of parenting stress (OR = 1.45, CI 95% 1.23; 1.72) were also positively associated with the presence of chronic pain in the child. Maternal depression, however, was not significantly related to chronic pain at T2.

## 5.3 Determinants of severe chronic pain

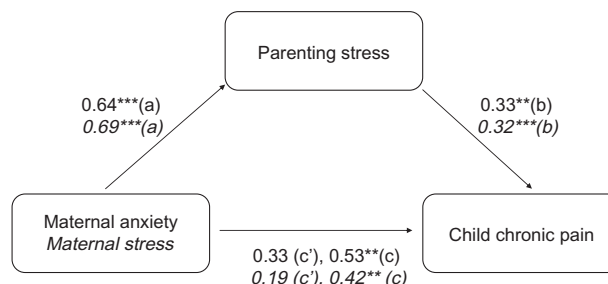
The same separate logistic regression analyses as above were run, but this time with severe chronic pain at T2 as the dependent variable, thus comparing children with severe chronic pain to children with no pain. The results show (Table 3) that the same determinants are of influence: maternal anxiety (OR = 1.85; CI 95% 1.20; 2.83), maternal stress (OR = 1.57; CI 95% 1.40; 2.17) and parenting stress (OR = 1.47; CI 95% 1.22; 1.77) were all positively related to the child's chronic pain. Maternal depression did not influence child severe chronic pain at T2. To summarize, mothers with high levels of anxiety, stress or parenting stress at T1 were more likely to have a child with chronic pain or severe chronic pain at T2 than those with low levels.

## 5.4 Mediation

The first conditions of the mediation analysis, which are the significant relationship of path *a*, *b* and *c* were met: with *a* being the relationship between maternal anxiety and parenting stress ( $a = 0.64$ ,  $p = 0.00$ ); with *b* being the relationship between parenting stress and child chronic pain ( $b = 0.33$ ,  $p = 0.00$ ); and *c* being the

relationship between maternal anxiety and child chronic pain ( $c = 0.53$ ,  $p = 0.009$ ). The bootstrap results indicated that the total effect of maternal anxiety on child chronic pain (total effect = 0.53,  $p = 0.009$ ) became non-significant when parenting stress was included as a mediator (0.33,  $p = 0.13$ , Fig. 1). These relationships were controlled for pain at T1 and child sex in the analyses. Furthermore, the analyses showed that the total indirect effect (i.e., the difference between the total and direct effects) of maternal anxiety on the presence of the chronic pain of the child through parenting stress was significant with a point estimate of 0.21 and a 95% bootstrap confidence interval of 0.096–0.351. Therefore, parenting stress mediated the association between maternal anxiety and child chronic pain.

A similar procedure with maternal stress at the independent variable showed that the first conditions of the mediation analysis, which are the significant relationship of path *a*, *b* and *c* were met: with *a* being the relationship between maternal stress and parenting stress ( $a = 0.69$ ,  $p = 0.00$ ); with *b* being the



**Figure 1** Results of mediation analyses, represented as unstandardized regression coefficients. Path values represent unstandardized regression coefficients. The value by *c* represents the total effect of maternal anxiety on child chronic pain before including the mediator (parenting stress). The value by *c'* represents the direct effect of maternal anxiety on child chronic pain after including the mediator. Information in italics represents the results for the mediation analysis with maternal stress at the independent variable. \* $p < .05$ . \*\* $p < .01$ . \*\*\* $p < 0.01$ .

relationship between parenting stress and child chronic pain ( $b = 0.32$ ,  $p = 0.00$ ); and  $c$  being the relationship between maternal stress and child chronic pain ( $c = 0.42$ ,  $p = 0.005$ ). The total effect of maternal stress on child chronic pain (total effect =  $0.42$ ,  $p = 0.005$ ) became nonsignificant when parenting stress was included as a mediator ( $0.19$ ,  $p = 0.26$ ). The analyses showed that the total indirect effect of maternal stress on child chronic pain through parenting stress was significant with a point estimate of  $0.22$  and a 95% bootstrap confidence interval of  $0.078$ – $0.356$ . Again, parenting stress mediated the association between maternal stress and child chronic pain.

### 5.5 Mediation with severe chronic pain as outcome variable

All mediation analyses were run again with severe chronic pain as the outcome measure ( $Y$ ). Results show that for maternal anxiety as the independent variable relationships  $a$  ( $0.64$ ,  $p = 0.00$ ),  $b$  ( $0.32$ ,  $p = 0.00$ ) and  $c$  ( $0.61$ ,  $p = 0.00$ ) were significant. The total effect ( $c$ ) of maternal anxiety on child severe chronic pain became non-significant when parenting stress was included as a mediator ( $0.41$ ,  $p = 0.08$ ). The total indirect effect of maternal anxiety on child severe chronic pain through parenting stress was significant with a point estimate of  $0.21$  and a 95% bootstrap confidence interval of  $0.075$ – $0.358$ . Again, parenting stress mediated the association between maternal anxiety and child severe chronic pain.

Results show that for maternal stress as the independent variable relationships  $a$  ( $0.69$ ,  $p = 0.00$ ),  $b$  ( $0.31$ ,  $p = 0.00$ ) and  $c$  ( $0.45$ ,  $p = 0.00$ ) were also significant. The total effect ( $c$ ) of maternal stress on child severe chronic pain became non-significant when parenting stress was included as a mediator ( $0.22$ ,  $p = 0.22$ ). The total indirect effect of maternal stress on child severe chronic pain through parenting stress was significant with a point estimate of  $0.22$  and a 95% bootstrap confidence interval of  $0.063$ – $0.369$ . In summary, the mediation analyses show that maternal anxiety and maternal stress are both mediated via parenting stress to determine the presence of chronic pain, as well as severe chronic pain.

## 6. Discussion

The results of this study show that maternal vulnerability plays a role in the presence of adolescent chronic pain in a community sample. Both maternal anxiety and maternal stress at T1 were determinants of child chronic pain at T2. The subgroup analyses

with adolescents with severe chronic pain show similar relationships in terms of predictors. This is not surprising since the majority of adolescents with chronic pain in our sample actually experienced severe chronic pain (77%). The current findings corroborate findings with regard to maternal anxiety that have been found in studies on clinical samples (Garber et al., 1990; Walker and Greene, 1989). In addition, this finding is in line with a large cohort study by Ramchandani and colleagues (2005, 2006), which showed that maternal anxiety was an important determinant of chronic abdominal pain in young children.

Our study was the first to investigate parenting stress as a determinant of adolescent chronic pain. First of all, we found that higher levels of parenting stress at T1 were related to the presence of chronic pain at T2. Furthermore the mediational bootstrapping analyses showed that parenting stress mediated the relationship between maternal anxiety and chronic pain, and the relationship between maternal stress and chronic pain. This is the first step towards investigating the pathways of influence when it comes to maternal vulnerability. Previous research has focused on parenting stress in chronic pain in children, but it was investigated as a consequence of chronic pain, rather than a possible determinant (Eccleston et al., 2004). Future research should focus on the influence of parenting stress on actual parenting behaviour, in the context of pain, which may in turn be influencing how the child feels and behaves.

There are several mechanisms by which maternal vulnerability can influence chronic pain in children. First, it may present an indication of expressing stress or anxiety through physical symptoms. Ramchandani and colleagues (2005, 2006) suggested that the link between maternal anxiety and abdominal pain in very young children may indicate that the pain is an expression of anxiety in the child. Second, social learning processes (Bandura, 1977; Craig, 1983; Levy et al., 2007) such as reinforcement and modelling may influence this relationship. Previous research has shown that parental reinforcement, such as paying attention to the pain and reassuring the child, influenced child symptoms (Walker et al., 1993; Whitehead et al., 1994; Wolff et al., 2009). Higher levels of parenting stress are an indication that parenting difficulties exist, which may influence parenting behaviour, such as paying too much attention to the child's expressions of pain. Modeling could be involved too, in cases where the mother has physical problems which are regularly expressed in the presence of her child. It is known that while chronic pain may be unexplained in its origin, maternal physical



factors may be of influence (Stanford et al., 2008). However, this remains speculation, as in the current study maternal physical factors, such as physical symptoms or chronic pain, were not investigated.

The present study has shown that in a community sample of just over 2000 adolescents aged between 13 and 15 years, approximately 13% report chronic pain, and almost 10% report experiencing severe chronic pain. While the occurrence of chronic pain in community samples has previously been found to be higher (up to 30% in Perquin et al., 2000), the number of adolescents with severe chronic pain (9.9%) is almost comparable. This might be explained by the fact that previous studies were focused on pain, whereas in our study the questions on pain were imbedded in a questionnaire that covered many aspects of the adolescent's life. The specific focus of a study may lead to bias in the subject to answer accordingly, which may explain the lower figures we found in comparison to previous studies (Swartz, 1999). This explanation is supported by the comparable figures of those with severe chronic pain, as here the pain itself is supposed to be a dominant factor in the report. Participants tended to suffer mostly from headache, back pain or leg pain, as has also been shown previously.

There are several limitations to this study. Firstly, even if the data represent a longitudinal investigation we cannot be completely sure of cause and effect relationships, since chronic pain was not measured at both time points. Rather, a rough approximation of pain at T1 was used to try to control for the influence of pain at T1. It is possible that the presence of chronic pain has led to higher levels of parenting stress, resulting from the parent having to deal with an adolescent with chronic pain, with parents feeling overwhelmed (Eccleston et al., 2004). Secondly, maternal physical factors, such as physical symptoms or chronic pain, were not examined. Thirdly, no questions were asked regarding a possible somatic cause of the pain the adolescents experienced. Fourthly, the results of the mediation analyses should be interpreted cautiously, as maternal anxiety, stress, depression and parenting stress were all measured at T1. In the mediation analyses maternal anxiety was mediated through parenting stress to influence child chronic pain. Ideally, one would have measured maternal anxiety and parenting stress at different time points, to strengthen the findings of this pathway. Finally, the role of the father has not been examined in this study, as a potential source of influence on chronic pain.

The strength of the study lies in the large number of adolescents questioned, the longitudinal nature and the elaborate questions posed to them regarding pain.

The present findings are very promising and warrant further research. Future studies should thoroughly explore additional factors relating to maternal vulnerability, such as actual parenting behaviour, since they may provide insight into different mechanisms leading to chronic pain. Maternal behaviour should be investigated more extensively over time as well, since resolving this lends itself to intervention, and the role of the father should also be explored. In addition, parenting stress should be looked into in younger children with chronic pain, since the influence of maternal factors in younger children is even greater. Concerning its potential for clinical practice, the findings suggest that interventions to diminish anxiety complaints and parenting stress in the mother may prevent chronic pain in later phases of childhood.

In conclusion, the current study has shown that the presence of chronic pain in adolescents is probably determined by aspects of maternal vulnerability. In addition, the influence of these determinants was mediated by parenting stress. Focusing on maternal vulnerability, parenting stress and actual behaviour can offer more insight into the pathways of development of chronic pain in children.

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